# Management of Female Hypospadias

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## Abbreviation

UTI Urinary Tract Infection

## 16.1 Introduction

Hypospadias in females is a rare but well-defined congenital anomaly due to arrest of the development of the sinus urogenitalis. The term "hypospadias feminis" means a congenital false urethral opening/fissure of the posterior (lower) wall of the urethra in the anterior vaginal wall proximal to the hymenal ring [1]. Male hypospadias is more common, and the male to female ratio reported is 150:1. Female hypospadias, however, seems to have attracted much less attention and many cases go undiagnosed, and most of these are case reports only. Nevertheless, when the patients were evaluated for incontinence, the incidence was much higher than reported. In the study by Hoebeke et al. (1999), 288 girls were referred for correlation between the functional voiding disorders and the meatal anomaly. Eighty-eight of them presented with meatal anomalies (24 hypospadias and 64 covered hypospadias) [2]. In another study of 12,739 patients evaluated in Neuro-urology department, 131 patients met the inclusion criteria, and 18 (13.74%) were diagnosed as cases of hypospadias [3]. This shows that the anomaly is underreported. Though the diagnosis and management of the female hypospadias are simple, the condition is easily overlooked as physicians are unaware of female hypospadias. Usually, it becomes significant only as a result of difficulty during catheterization because of the inability to localize the meatus. All variants of female hypospadias should be surgically corrected by transposition of the external opening of the urethra from the vagina on the perineum under the clitoris to cure the chronic urethritis, cystitis and vulvovaginitis and chronic/recurrent UTI with hypospadias.

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## 16.2 Embryology

True hypospadias with failure of fusion of the urethral folds cannot occur in females since that portion of the urethra does not develop. However, the urethral meatus opens high up in the anterior vaginal wall and assumes a hypospadiac position. The etiology of this condition remains obscure. Embryologically, it is believed to be due to abnormal development of the urogenital sinus in distal hypospadias, or a lack of differentiation of Wolff's tissue, when it is proximal. The lower part of the urethra and the vagina has a common origin from the lower part of the urogenital sinus, and failure of the normal development of this structure may explain hypospadias formation. In an experimental study (Miyagawa et al.) in the mice, hypospadias was induced by injecting diethylstilbestrol [4]. Doses response analysis indicated that 0.03 g diethylstilbestrol for 5 days is the lowest known critical dose for hypospadias induction. The authors have shown that diethylstilbestrol-induced female hypospadias onset may primarily be the result of changes in developing dorsal urethral epithelial cell apoptotic and proliferative activity. The location of diethylstilbestrol-induced hypospadias formation is dependent on age at the time of exposure. Sometimes a part of hymen covers the hypospadiac opening and is called webbed hypospadias. The urinary stream in these cases may be directed anteriorly.

Other congenital anomalies of the urogenital septum like bicornuate uterus, vaginal septum, and vaginal atresia may be associated with hypospadias. Urethral abnormalities like urethral atresia, urethral stenosis, urethral duplication, and dorsal urethral epispadias may also be associated with female hypospadias.

## 16.3 Classification

### 16.3.1 Congenital

Most of the time, the urethral opening is in anterior vaginal wall proximal to introitus; the patient is vaginal voider and presents with incontinence (Fig. 16.1a, b). But the hypospadiac opening is often partially covered by hymen, and the urinary stream is directed anteriorly. The web cover may be partial (Fig. 16.2a) or complete (Fig. 16.2b).

### 16.3.2 Acquired Hypospadias

The cause of female hypospadias in the older age group may not be congenital. The urethral meatus in such cases may become drawn up due to fibrosis after atrophic vaginitis or surgery on the urethra (Fig. 16.3a, b). Sometimes the patient may be subjected to female genital mutilation during early childhood, which is a tradition practiced in West African Yoruba tribe, and this may cause the urethral opening in hypospadiac location [5]. The long-term catheterization and poor catheter management in neurogenic bladder or old bedridden patient may lead to ventral urethra erosion and pooling the meatus high up in the vagina.

### 16.3.3 Blum Classification [6]

He divided these cases into three groups:

1. A longitudinal communication between the posterior wall of the urethra and the anterior wall of the vagina.
2. A persistent urogenital sinus, where the vagina enters into the urethra, but the hymen lies deep in the urogenital sinus.
3. The urethra opening into the vagina proximal to a normal hymen.

### 16.3.4 Solov'ev Classification [7]

1. Vestibular (partial)
2. Vestibulovaginal (subtotal)
3. Vaginal (total)

### 16.3.5 Clinical Classification

The clinical classification consists of complete (type I) and incomplete (type II); urethral subtypes are II-a, short wide, and II-b, standard urethral diameter.

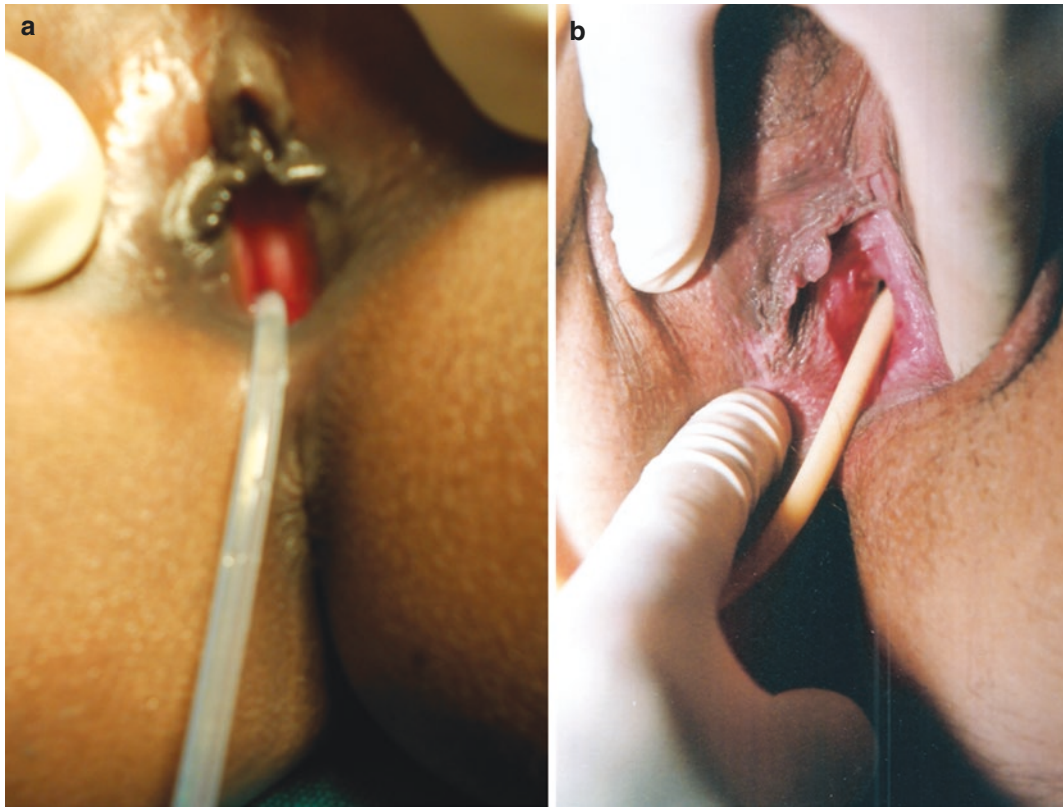


**Fig. 16.1** Congenital female hypospadias in childhood and adult. (**a**) Congenital hypospadias with a catheter in hypospadiac opening high in the vagina in a child. (**b**) Congenital hypospadias with a catheter in hypospadiac opening high in the vagina in an adult

### 16.3.6 Derevianko Classification of Anatomic Variants of Female Hypospadias [8]

1. Low vaginal ectopia: Ectopia of the external urethral opening still outside the introitus
2. High vaginal ectopia of the external opening of the urethra lying inside the introitus
3. Urovaginal (vesicovaginal) fusion of the neck of the urinary bladder with vagina accompanied with enuresis, whole urethra is opened
4. Ectopia of the external urethral opening in the urogenital sinus, i.e. urogenital sinus in females
5. Any of the above variants of female hypospadias in combination with false or true hermaphroditism.

## 16.4 Presentation

Most of the time, female hypospadias goes unnoticed. It needs a high index of suspicion to diagnose the female hypospadias. Female hypospadias should be suspected in the following conditions:

1. Urinary incontinence
2. Recurrent UTI
3. Chronic UTI
4. Urethral syndrome
5. Dysfunctional voiding
6. Dyspareunia
7. Urethritis, cystitis, and vulvovaginitis
8. Cervico-vaginitis, endometritis, and secondary infertility

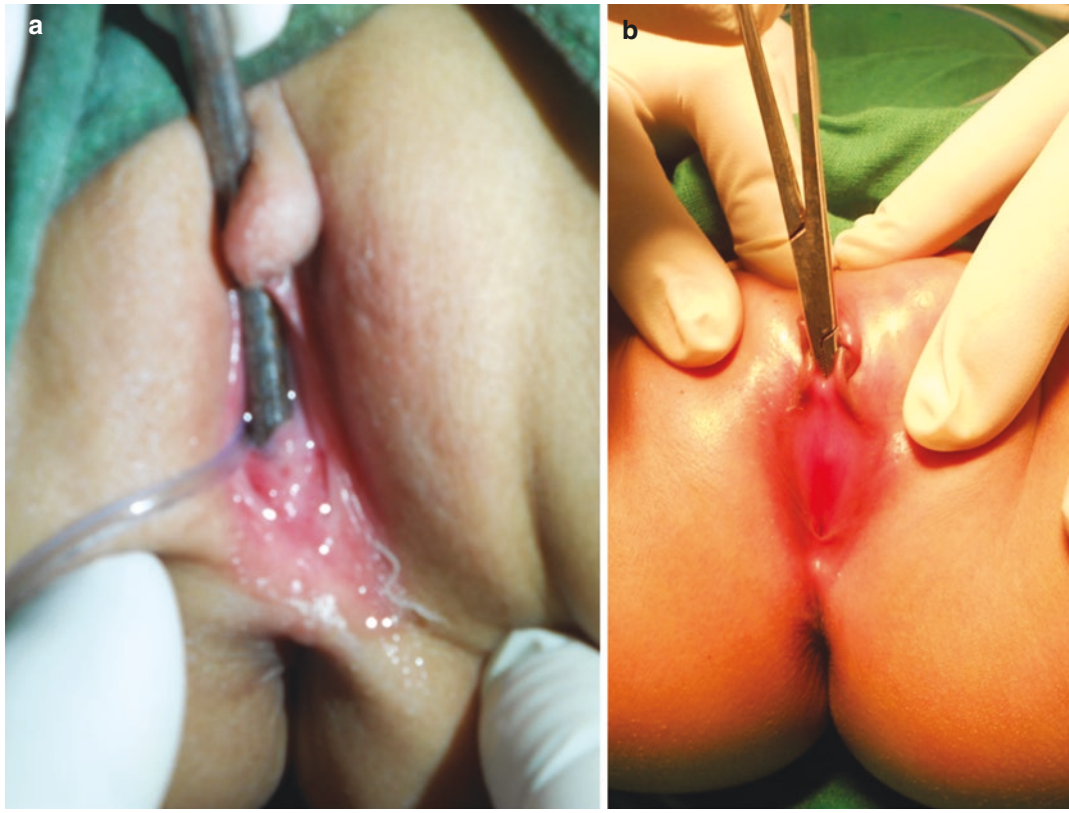


**Fig. 16.2** Congenital webbed female hypospadias. (**a**) Partial/incomplete webbing with a catheter in hypospadiac opening. (**b**) Complete webbing with imperforate hymen Hemostat showing the common opening just below the clitoris

A urethra of standard caliber without meatal stenosis may be asymptomatic, coming to light only when an attempt is made to catheterize the patient. Incontinence of urine is an important sign of female hypospadias. Incontinence can be true, determined by a short urethra on which the pelvic muscular complex could not act at best and can be pseudo-incontinence or post micturition incontinence, imperfect control caused by vaginal voiding. They can have urethral syndrome (frequency, dysuria, urgency), recurrent UTI, and dyspareunia presenting when a sexually active life has started. If the urethra is narrow, the presentation is more likely to have signs of urinary outflow obstruction, distended urinary tract, urinary tract infection, obstructive nephropathy, and hypertension. Webbed hypospadias may present with the direction of the stream towards the face. The web may cover the meatus completely with imperforate hymen or may cover the meatus partially. So important observation of the parents may be that baby soils the chest and face by the urinary stream, or the baby does not pass urine in stream.

## 16.5 Diagnosis

Diagnosis is usually by clinical examination to locate the meatus, and hardly any investigation is required. Inability to locate the meatus for catheterization should raise the suspicion of the hypospadias. Failed catheterization in a female is a cardinal sign in the diagnosis of hypospadias. Examination of the external genitalia of the baby may show leakage of urine from the vagina with an absence of urethral opening (Fig. 16.4a, b).

Per speculum examination is required to locate the meatus in the anterior vaginal wall in adults (Fig. 16.3b). Intravenous Urogram or CT Urogram is advisable to rule out the upper tract

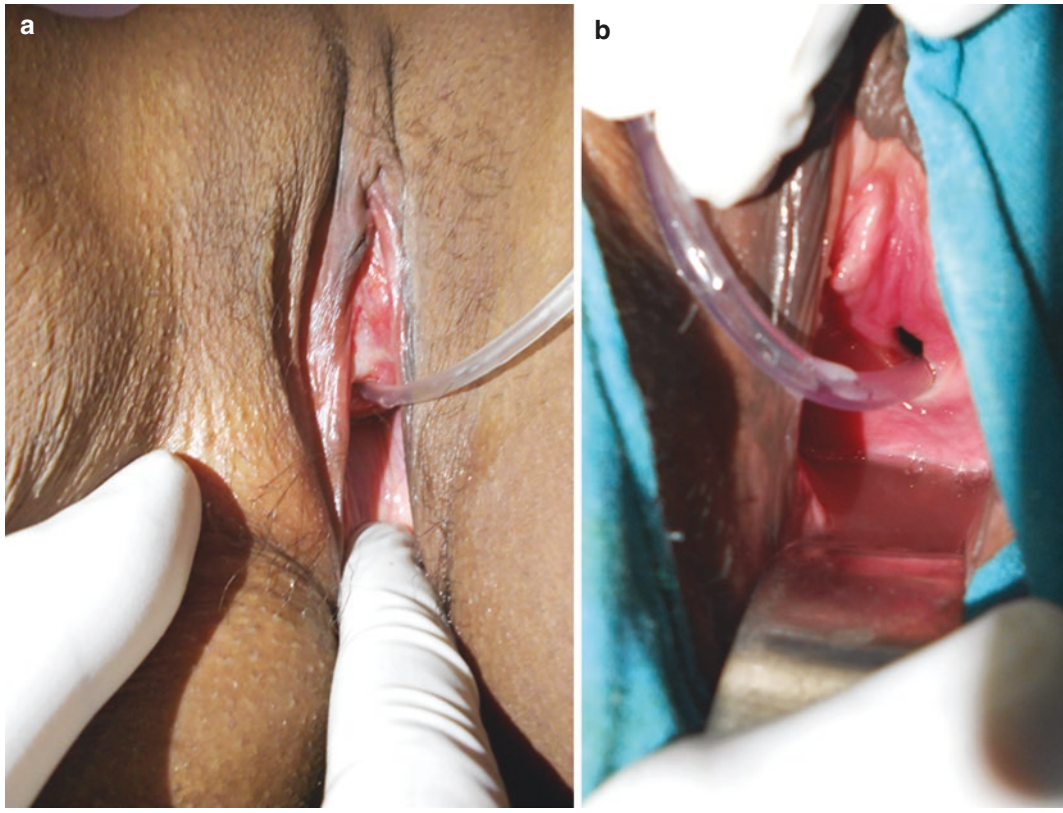


**Fig. 16.3** Acquired female hypospadias. (**a**) Urethral opening high in the vagina with atrophic changes. (**b**) Urethral very opening high in the vagina just at the bladder neck

abnormalities. Micturating cystourethrogram may give of vaginal spillage of dye but seems to be an academic exercise. Cystoscopy or catheterization clichés the diagnosis, showing short urethra lying urethral opening high in the vagina (Fig. 16.1).

## 16.6 Female Hypospadias and Dysfunctional Voiding

Female hypospadias is rare, and many a time patient present as a urethral syndrome. A majority of the women with the urethral syndrome have had dysfunctional voiding since childhood. This suggests a causal association between dysfunctional voiding and minimal meatal deformities. A variety of hypotheses have been proposed to explain this association [2]. The bulbocavernosus reflex that is usually absent during voiding. However, it is generally elicited by genital stimulation. In patients with anterior deflection of the urinary stream, the stream passes the clitoris and can stimulate the bulbocavernosus reflex, which in turn can initiate sphincter activity during voiding. Strong vaginal voiding in cases of female hypospadias could provoke the same reflex. In neurogenic bladder with detrusor sphincter dyssynergia, where the neurological lesion deletes normal inhibition of the bulbocavernosus reflex during voiding; it is similar to the mechanism as seen in female hypospadias. In girls with dysfunctional voiding, this inhibition might not be obtained because of the minimal anatomic deformity. The only defiance against voiding over the rim of the toilet may be a bent posture, that precludes good relaxation of the pelvic floor muscles during voiding, thus creating a functional obstruction during voiding in girls with anterior deflection of the urinary stream. And in girls with hypospadias, extreme vaginal voiding can cause further urine loss after voiding. To prevent this,

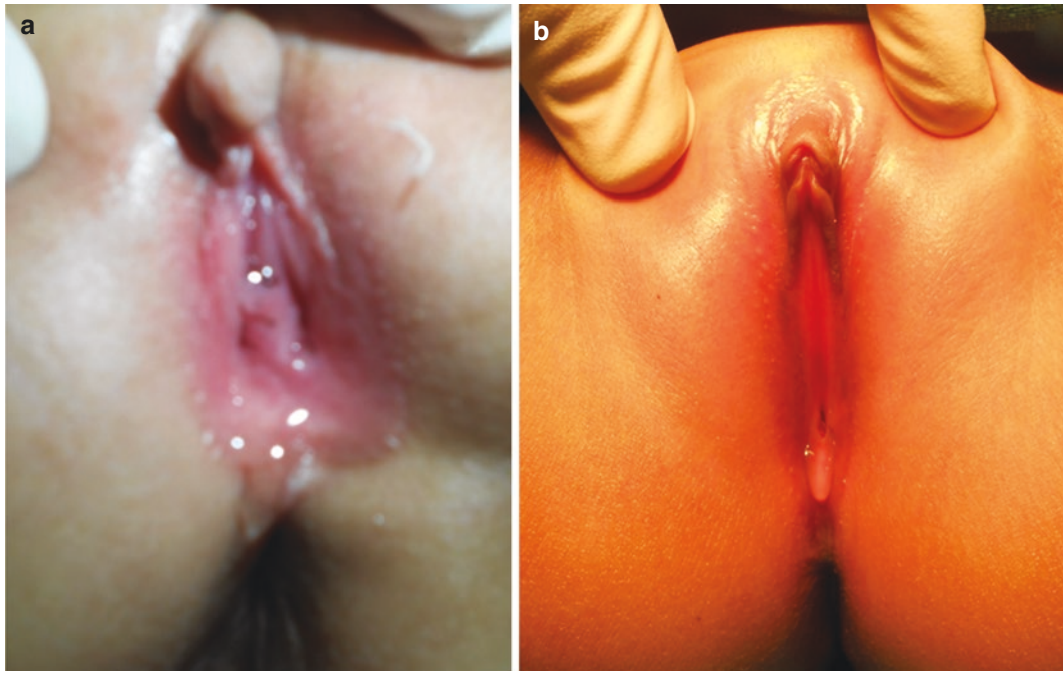


**Fig. 16.4** (**a** and **b**) Congenital female hypospadias with urinary leakage and absent urethral meatus in childhood

the girls may contract their sphincter intensely several times a day, which might be responsible for sphincter hypertrophy, finally resulting in dysfunctional voiding. Vaginal filling during voiding can even stimulate the bulbocavernosus reflex.

## 16.7 Urinary Incontinence in Female Hypospadias

Patients of female hypospadias may have stress incontinence, pseudo-incontinence and even genuine incontinence. Patients are the vaginal voider, so such patients never void in-stream which may be elicited on detailed interrogation while taking history. Urine pooled in vagina later soils the undergarments giving the impression in incontinence, but that is pseudo-incontinence (Fig. 16.4a, b). Many patients of female hypospadias present as stress incontinence after vaginal delivery and sometimes with total incontinence. The total incontinence in severe hypospadias is due to the location of meatus at bladder neck making it incompetent. Genuine incontinence and stress are more common in urethra-vesical and high vaginal hypospadias.

## 16.8 Vaginal Stones

Vaginal stone with hypospadias is a rare condition; only one case has been reported. Urethral opening high in anterior vaginal wall proximal to hymen will lead to voiding in the vaginal cavity, so much so that patients never voids in a stream. With an intact hymen, there is stagnation of urine in the vagina which may lead encrustation in the vaginal cavity. If a patient is not managed in time, then there may be stone formation and stone may take the shape of the vaginal cavity (Fig. 16.5). Neglected patients with a vagina full of rocks may have coital difficulty and dyspareunia. The vaginal examination may reveal a stone palpable in the vagina and if it enlarges in size and may protrude out of the introitus. We came across one such case who was married and presented with dyspareunia [1].

## 16.9 Infertility

Female hypospadias may be an unusual cause of infertility. Voiding urine in the vaginal cavity and stagnation of urine in the vagina adversely affect

**Fig. 16.5** Vaginal cavity shaped stones removed from the vagina (copy from Bhat et al. [1] with permission)

the normal vaginal flora and cause infection in the vaginal cavity. Presence of infected urine in the vagina spreads it to cervix and uterus. The repeated cervicovaginal infections may even lead to chronic or intermittent endometritis and infertility. Secondly, urine washes the vaginal secretion, which will lead to vaginal dryness leading to dyspareunia and contributes to the risk of infertility. Vaginal flap urethroplasty prevents urination into the vaginal cavity and vaginal infection. Control of cervico-vagino-uterine infection and vaginal lubrication with normal secretions reduces the dyspareunia, which helps cure secondary infertility.

## 16.10 Management

Surgery is the treatment of choice in symptomatic female hypospadias. Both in primary and secondary hypospadias, urethral reconstruction should be done to prevent the sequelae and complication of the hypospadias. Hymenotomy in infants and children may cure the recurrent urinary tract infection.

### 16.10.1 Vaginal Flap Urethroplasty

A vaginal flap urethroplasty is performed, keeping the patient in a frog-leg position/lithotomy position under general anesthesia. An inverted U-shaped incision is given around the urethral meatus and then extended into the anterior vaginal wall after putting in the catheter (Fig. 16.6a, b, c). Anterior vaginal wall mucosal flaps are raised on both sides of the incision (Fig. 16.6d, e). A urethral tube is constructed by tubularization of vaginal flaps to bring the external meatus to the base of the clitoris over an adequate size catheter. The incision is extended deep into the muscle layer of the anterior vaginal wall, and this surrounding tissue along the muscle is stitched over the newly constructed urethral tube as a second layer. Vaginal mucosal flaps are dissected on both sides of the incision and sutured with interposition of a pedicled adipose tissue flap (Fig. 16.6f). The neo-meatus is reconstructed between the introitus and the clitoris (Fig. 16.6g).

Figure 16.7 The diagrammatic presentation of vaginal flap urethroplasty. In most of the simple cases of the vaginal flap, urethroplasty is sufficient to control the symptoms. Still, surgical management of cases of female hypospadias with stricture may be technically more difficult, as the stricture would further shorten the already short hypospadiac urethra. A combination of urethrolysis, urethral transposition, and urethroplasty, or some other cumbersome amalgam of procedures may be needed for satisfactory resolution of the symptoms.

### 16.10.2 Hymenotomy/Web Incision

In cases with a meatal mucosal web, the web is incised longitudinally and closed transversely with three interrupted polyglactin 6/0 sutures, which corrects the direction of the urinary stream (Fig. 16.8a). Partial hymenotomy/incision in the hymen is given to open up the meatus (Fig. 16.8b) which corrects the urinary stream pointing forward, and vaginal collection of urine is corrected (Fig. 16.8c). If later on problem persists, then vaginal flap urethroplasty may be required.

## 16.11 Our Experience and a Brief Review

We published four cases of female hypospadias; one of them having vaginal stones in 2010 [1].

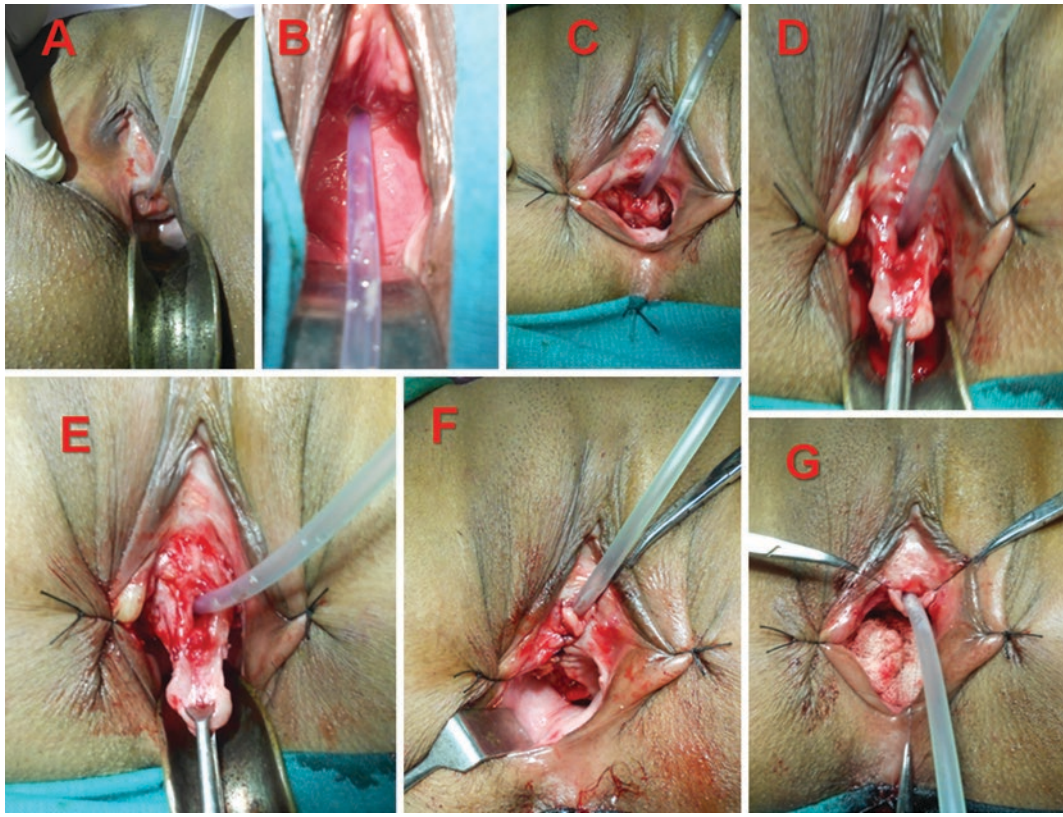


**Fig. 16.6** Vaginal flap urethroplasty in acquired hypospadias. (**a**) and (**b**) Atrophic vagina with urethral opening high in the vagina almost at the bladder neck. (**c**) Inverted U-shaped incision in the anterior vaginal wall. (**d**) Vaginal wall flap raised. (**e**) Similar flap marked dorsal to the meatus. (**f**) Both sutured together to bring the meatus at the usual location and vaginal wall sutured over it. (**g**) Vaginal wall closure and wide normal located meatus

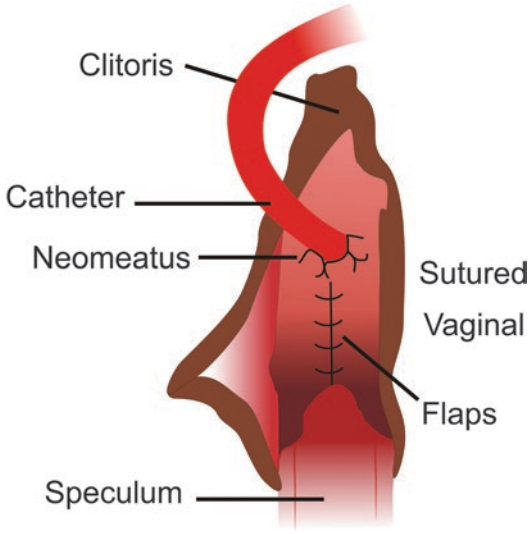


**Fig. 16.7** Diagrammatic representation of vaginal flap urethroplasty

Later on, we retrospectively reviewed of cases sheets of females presenting with recurrent UTI, chronic UTI, incontinence, and vaginal discharge from 2012 to 2019. During this period, we had a total of nine (five congenital and four acquired) cases of female hypospadias. Clinical history details were obtained, and thorough clinical examinations, urinary catheterization, and cystoscopy were done. Genitalia examination, observing leakage of urine from vagina, catheterization, and/or cystoscopy clinched the diagnosis. Management was done on a case to case-based, and results were recorded. Age of the patients ranged from 6 months to 25 years in congenital and 55 years to 68 years in acquired cases with a mean age of 29.6 years—all of the patients presented with recurrent UTI and recurrent UTI. One of the patients had stream directed towards the abdomen, and two had leak-

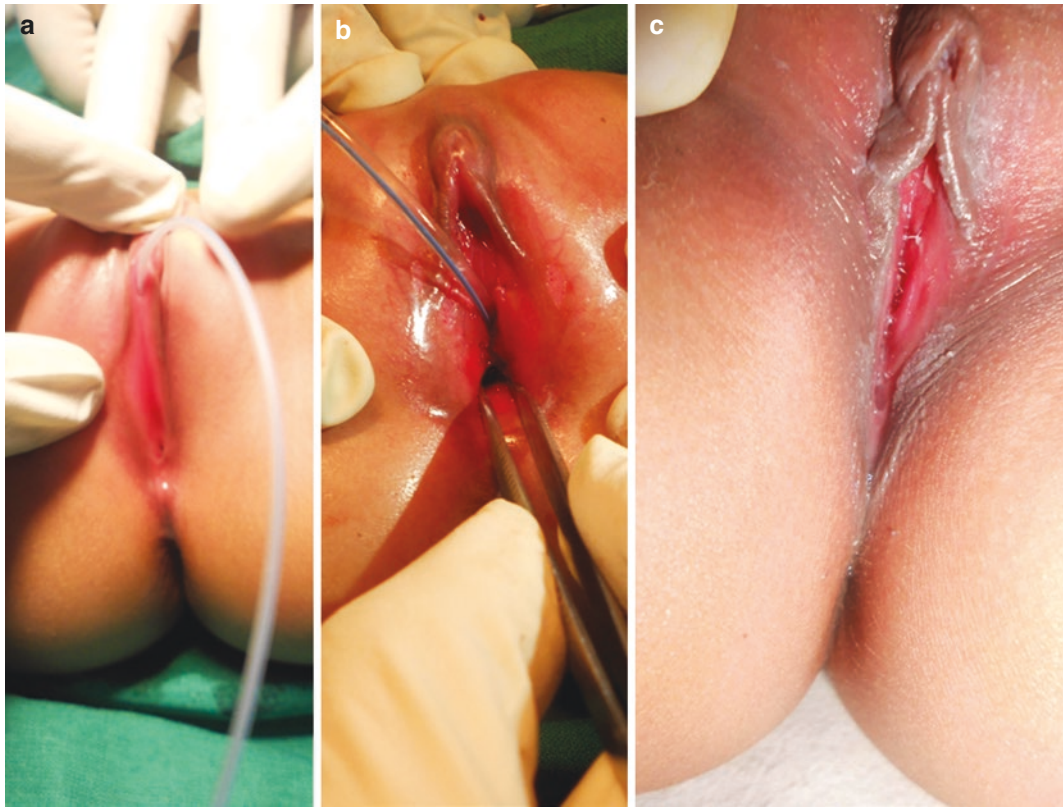


**Fig. 16.8** Hymenotomy in hypospadias with imperforate hymen in a patient with a urinary stream directed to face. (**a**) Intact hymen with an opening just below the clitoris. (**b**) Hymen incised to open up the urethral meatus and catheter in the meatus. (**c**) Postoperative, after 3 weeks, meatus still high in the anterior vaginal wall

age of urine from the vagina. Two of the patients had urinary incontinence since birth and recurrent attacks of UTI. The fourth patient complained of dyspareunia. The diagnosis of female hypospadias was made by catheterization, culdoscopy, voiding cystourethrogram, and pan-endoscopy. The patient was put in a frog-leg position, and a peri-urethral vaginal flap urethroplasty was done in three layers. Excellent results were seen in three cases. Two of the patients were managed by partial hymenotomy, the release of the fused membrane was done in two, and supra-pubic cystostomy had to be performed in two cases with urethral erosion. Postoperative period was uneventful, and sterile urine culture was obtained up to 24–36 months of follow-up. Patients of partial hymenotomy are still under follow-up for any problem requiring urethroplasty.

Review of the literature shows that most of the cases are case reports, go undiagnosed and are diagnosed secondary to morbid complication. The studies conducted for evaluation of the patients of incontinence, recurrent UTI, and the incidence of hypospadias in girls with abnormal urodynamic ; the incidence has been as high as 15% of hypospadias which shows that anomaly is not as rare as has been reported. Diagnosis in most of the cases is made either in adulthood with some complication of hypospadias is again an important fact. Urinary incontinence is the commonest symptom, followed by recurrent/chronic UTI. Commonest complication observed was a renal failure due to late diagnosis and treatment. Vaginal pooling of urine may lead to a rare complication of vaginal stones. Once the diagnosis has been confirmed, the outcome of surgery and management had been successful. Salient feature and results of the reported cases in the literature are shown in Table 16.1.

**Table 16.1** Reported cases management and results

| S. N. | Authors | Year | No of cases | Age (Years) | Presentation | Complication / Sequalae | Procedure / management | Result |
|---|---|---|---|---|---|---|---|---|
| 1. | Bhat A et al [1] | 2010 | 4 | 1. 18-65 Years | incontinence of urine, recurrent UTIs since childhood and dyspareunia Inability to Catheterize. Retention of urine | Vaginal stone | Vaginal flap urethroplasty<br>Urethral dilatation and catheterization | Good |
| 2 | OEBEKE P [2] | 1999 | 24 Hypospadias64 covered hypospadias | | Urinary incontinence , Neurogenic bladder | - | – | – |
| 3 | Ronzoni G etbal [9] | 2001 | 32 | Mean age 22 range 16-38 | diurnal nocturnal frequency, dysuria, urgency, suprapubic pain and sometimes urge incontinence. Failed medical treatment | - | Urethral translocation | Good |
| 4 | Van Bogaert L J [10] | 1992 | 6 | Adult females | Urethral Syndrome Failed medical treatment | -- | Tranlocation of meatus | Good |
| 5 | Ayed M<br>et al. [11] | 1995 | 3 | Adult | Urinary incontinence<br>Vaginal Atresia | | Vaginal Flap urethroplasty | Re- operation -1 |
| 6 | Antolak et al[12] | 1969 | 2 | 2 Years<br>3 Months | Urethral Calibration<br>Vaginal flap urethroplasty Urethral calibration | Pooling of urine in vagina | Vaginal flap urethroplasty | Nil |
| 7 | Bouty A et al [13] | 2016 | 2 | < 5 Years | Associated with urethral duplication | - | 1.Dorsal urethra resection & Hypospadiac urethral dilatation.<br>2.Partial urogenital sinus mobilization | Good |
| 8 | D'Cunha AR, et al [14]. | 2016 | 1 | 1. 4 Years | Straining during micturition , Ambiguous genitalia | Renal failure | Vesicostomy | Renal function improved |
| 8 | Ravichandran R et al [15] | 1995 | 2 | 3 and 9 Years | Incontinence of urine | Neurogenic bladder | Mitrofanoff operation and CIC | Good |
| 9 | Tsujimoto Y et al. [16] | 1984 | 1 | 24 Years | Urinary frequency and incontinence | Left NFK | Bladder neck and urethral narrowing & suspension bladder neck | Good |
| 10 | Sarin YK Kumar P {17} | 2019 | 1 | 6 Years | Urinary incontinence | - | external urethral lengthening | Satisfactory |
| 11 | Patil N A et al [18] | | 1 | 7 Years | Urinary incontinence | Small capacity bladder | Vaginal Flap urethroplasty | Good |
| 12 | Lima M. et al [19] | 2018 | 1 | 5 Years | Recurrent UTI and VU reflux | Vu reflux | -Vaginal flap urethroplasty and ureteric reimplantation | Good |
| 13 | Niyagi T et al<br>[20] | 2020 | 1 | 21 Years | secondary infertility, dyspareunia, and urge symptom | Secondary<br>infertility | Vaginal flap urethroplasty | Good |
| 15. | Mildburger H et al [21] | | 1 | 1 Years | With supra-pubic diversion due to wrong diagnosis of occult neurogenic outlet obstruction and diagnosed during investigation and un-diversion was done. | | Undiversion ,And catherization | Good |
| 16 | Gaurav Prakash et al [22] | 2016 | 1 | 11 Years | Chronic retention of urine with CRF<br>Failed per urethral catheterization | Chronic Renal Failure | Suprapubic Catheterization ,Antegrade cystoscopy and catheterization | Good |
| 17 | Bello OJ et al [23] | 2012 | 1 | 68 Years | Obstructive uropathy Stricture urethra CRF | Chronic Renal Failure | Urethral dilatation and CIC | Good |

## 16.12 Conclusions

A high index of suspicion is required to diagnose hypospadias, especially in patients presenting with an abnormal urinary stream, vaginal discharge, urinary incontinence, urethral syndrome, recurrent UTI, and chronic UTI. Senile atrophic vaginitis and urethral erosion and loss may be the cause of secondary hypospadias in patients on long-term catheterization. Absence of urethral opening with vaginal voiding or urinary dribble is a cardinal sign of hypospadias in a female child. Diagnosis is made during catheterization and/or lower tract endoscopy. Vaginal voiding leads to urinary stagnation in the vagina, causing urinary pseudo-incontinence and vaginal infection. A good outcome is expected by early diagnosis and management. Vaginal flap urethroplasty and approximation of peri-urethral smooth muscle help in continence by creating a pseudo-sphincter.

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